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Characterization and outcomes of gastroschisis in two institutions of a middle-income country in Cali, Colombia, between 2018 and 2022

Javier Torres-Muñoz[*1] , Luis Mauricio Figueroa Gutierrrez[1,2], Mariana Ángel Correa[3], Sofia Torres Figueroa[1,4], José Félix Ibarra Mera[5]

1. INSIDE Research Group. Department of Pediatrics of the Universidad del Valle, Hospital Universitario del Valle, Cali, Colombia. 2. Department of Surgery, Pediatric Surgery section of the Universidad del Valle, Hospital Universitario del Valle, Cali, Colombia. 3. Health Faculty, Medicine and Surgery Program, School of Medicine, Universidad del Valle, Cali, Colombia. 4. Faculty of Health Sciences, ICESI University, Cali, Colombia. 5. Faculty of Health Sciences of the Pontifical University Javeriana Cali, Colombia

Correspondence*: Javier Torres Muñoz, INSIDE Research Group. Department of Pediatrics of the Universidad del Valle, Hospital Universitario del Valle, Cali, Colombia. **E-mail**: javier.torres@correounivalle.edu.co

KEYWORDS

Gastroschisis, Abdominal wall, Neonatology, Congenital

ABSTRACT

Background: Gastroschisis is the most common congenital defect of the abdominal wall. Its incidence has increased by 2 to 10 times worldwide. However, morbidity and mortality rates vary between high-, medium-, and low-income countries.

Methods: This is an observational, cross-sectional study aimed at establishing the prevalence of gastroschisis in babies admitted to the intensive care units of the Clinica Versalles and the Hospital Universitario del Valle (HUV). Additionally, it is analytical, as it seeks to identify the clinical and surgical factors associated with the mortality of these neonates. Data were collected based on the information already recorded in the clinical histories of patients born or referred to these institutions between January 2018 and June 2022.

Results: Fifty-six (56) babies were evaluated, with a birth rate corresponding to 1/1,000 live births. Of these, 35.70% were born to mothers under 19 years old, 55.36% came from municipalities other than Cali, and 91.07% were born at a gestational age greater than 32 weeks. Additionally, 58.90% were girls, with a median weight of 2,167 g (range: 1,440–3,240 g). Among the evaluated babies, 25% presented with complicated gastroschisis, and 44.64% had other malformations. The association between mortality outcomes and clinical factors showed that invasive mechanical ventilation for longer than 7 days increased the probability of death by 7.94 times, sepsis by 8.35 times, pulmonary hypertension by 7.14 times, acute kidney injury by 8.87 times, and pneumonia by 7.14 times. The surgical factors significantly associated with mortality were abdominal compartment syndrome (13.67 times), staged closure (9.17 times), more than two surgical interventions (7.80 times), failure to achieve closure (30.71 times), post-surgical complications (5.29 times), and intestinal ischemia (43 times).

Conclusion: The rate of gastroschisis in this study is notably higher than that reported in other countries, with a substantial number of clinical and surgical complications significantly associated with mortality. Key factors influencing mortality include prolonged invasive mechanical ventilation, sepsis, pulmonary hypertension, acute kidney injury, and pneumonia. Surgical factors such as abdominal compartment syndrome, staged closure, multiple surgical interventions, failure to achieve closure, post-surgical complications, and intestinal ischemia also greatly increase the risk of death.

INTRODUCTION

Gastroschisis is a congenital abdominal wall defect characterized by paraumbilical intestinal herniation [1][2]. It is classified as simple when there are no intestinal complications and complex when intestinal atresia, necrosis, stenosis, perforation, or volvulus is present [3–5]. It represents the most common abdominal wall defect in neonates worldwide, with a rate of 1.1 to 5.1 per 10,000 live births and an increase in incidence of 2-10 times over the last three decades [6]. Gastroschisis disproportionately affects young mothers (< 20 years of age), with 70% of neonates with gastroschisis born to mothers under 25 years of age [7]. In most cases, pregnant women with gastroschisis have had good prenatal care [1].

Between 10 to 20% of gastroschisis cases have an associated anomaly, usually of the gastrointestinal tract [6][8], with 10% involving intestinal stenosis or atresia [1]. Left-sided gastroschisis is a rare condition that occurs mostly in females and is associated with a

higher incidence of extraintestinal anomalies [1]. Gastroschisis is more common in Caucasians of Northern Europe [9]. The incidence of intrauterine fetal death due to gastroschisis is 5% [1]. The mortality rate varies according to its classification, being higher in complex gastroschisis (16.67%) than in simple gastroschisis (2.18%) [1].

In studies conducted in Latin America, the incidence of gastroschisis has increased from 0.77/10,000 births to 10/10,000 [10][11]. Studies carried out at this center in Cali, Colombia between 2000 and 2008 reported an incidence of 5.15/10,000 births [12][13]. Therefore, one objective of this research was to understand the changes in the incidence of this malformation in our environment.

This study describes the clinical and surgical characteristics and the factors associated with mortality from gastroschisis in a low- and middle-income population over four years (2018-2022), aiming to identify the changes in the incidence of this malformation in our environment.

METHODS

Definitions

The term primary closure indicates the complete reduction of the defect with surgical closure of the abdominal wall on the first day of life, either with or without sutures [14]. Staged or delayed closure, contrary to primary closure refers to closure by stages after an initial surgical intervention where complete reduction of the protruded organs or abdominal wall closure is not immediately possible. Staged closure can involve the use of a silo (in cases of an irreducible defect) or a delayed abdominal wall closure, where the open abdomen is covered with a tight waterproof dressing, with or without a vacuum-assisted closure (VAC) system [15].

The decision between performing a staged closure or a primary closure is made by the surgeons based on the size of the abdominal cavity (visceral-abdominal disproportion) and the presence of hemodynamic instability once the organs are first reduced. Once abdominal wall closure can be performed without respiratory compromise, it is carried out in a subsequent surgical intervention.

Completion of enteral feeding requirements is considered achieved upon reaching 120 kilocalories per kilogram per day. Post-surgical complications include stenosis, the need for intestinal resection, obstruction, short-bowel syndrome, the requirement for an ostomy, evisceration, intestinal ischemia, or intestinal perforation [16].

Type of Study:

This is an observational, cross-sectional study aimed at establishing the prevalence of gastroschisis in babies admitted to the intensive care units of the Clinica Versalles and the Hospital Universitario del Valle (HUV). Additionally, it is analytical, as it seeks to identify the clinical and surgical factors associated with the mortality of these neonates. Data were collected based on the information already recorded in the clinical histories of patients born or referred to these institutions between January 2018 and June 2022.

Study Population

Neonates who were treated in the intensive care units (UCIN) at HUV and Clinica Versalles between January 2018 and June 2022.

Inclusion Criteria: Newborns diagnosed with gastroschisis, all of whom were born in the study institutions.

Exclusion Criteria: Newborns whose medical histories did not have the complete data required for the analysis of the study.

The sample size comprised the total number of babies born with this diagnosis during the four years included. The dependent variable, gastroschisis, was evaluated according to clinical, sociodemographic, and mortality outcomes.

Analysis of Data

The data used in this study were extracted from the patients' medical records and stored in a database using Epi Info software version 7.2.5.0, which was then exported to Stata software version 14® for statistical analysis. Initially, an exploratory data analysis was conducted, calculating measures of central tendency and dispersion according to the behavior of the quantitative variables. Qualitative and categorical variables were organized into frequency and percentage distributions.

For the bivariate analysis, the Shapiro-Wilk test was applied to the quantitative variables to evaluate normality. Hypothesis tests were applied to evaluate possible statistical relationships. For normally distributed variables, Student's t-test was used for two groups and ANOVA for more than two groups. For variables that were not normally distributed, the Wilcoxon test was used for two groups and the Kruskal-Wallis test for more than two groups. For categorical variables, the chi-square test or Fisher's exact test was used in cases where one of the variable categories had five or fewer data points. Finally, a logistic regression model was built to explore the associations of the odds ratios (OR) and their respective confidence intervals (95% CI) between the covariates and the variable of interest. Data analysis and processing were carried out using Stata version 14.

Ethical Statement

The study was conducted in accordance with the guidelines of the Declaration of Helsinki and was approved by the Institutional and Ethics Committee of the Hospital Universitario del Valle (code 051-2022), Clinica Versalles (code CV-PI-0105), and Universidad del Valle (code E040-022).

RESULTS

During the study period, 60 patients were born with a diagnosis of gastroschisis. Out of these, 56 were included in the study, as 4 did not have complete information. Among the included patients, 58.9% were female, with a median gestational age of 35 weeks (range: 30–38 weeks). The median birth weight was 2,167.5 grams (range: 1,440–3,240 grams). 87.5% had adequate prenatal care, defined as more than three prenatal visits, and 91% achieved a gestational age greater than 32 weeks. The median maternal age was 19.5 years, with 35.7% of mothers being under 19 years old. In the vast majority of cases, the diagnosis of gastroschisis was made prenatally (96.4%), mainly in the second trimester (63%).

39.3% (22 patients) presented intrauterine growth restriction, and 44.6% (25 patients) had other associated malformations. Of the sample, 73.2% (41 patients) were hospitalized in a public institution, and 26.8% (15 patients) in a private one. The birth was by cesarean section in 60.7% (34 cases). Complicated gastroschisis occurred in 25% (14 cases).

Table 1: Description of the demographic and clinical variables.

VARIABLES	Number and percentage or median
Female sex	33 (58.9%)
Gestational age at birth (median)	35 weeks (min. 30 – max. 38)
Weight at birth (median)	2,167.5 gr (1,440gr – 3,240gr)
Prenatal control (≥ 3 doctor's visits)	49 (87.5%)
Gestational age greater than 32 weeks	51 (91.07%)
From municipalities outside Cali	31 (55.36%)
From eastern Cali*	12 (50%)
Maternal age (median)	19.5 years old (min. 14, max. 34)
Maternal age < 19 years old	20 (35.7%)
Prenatal diagnosis	54 (96.4%)
Prenatal diagnosis during 1st trimester	8 (14.8%)
Prenatal diagnosis during 2nd trimester	34 (63.0%)
Prenatal diagnosis during 3rd trimester	12 (22.2%)
Intrauterine growth restriction	22 (39.3%)
Other congenital anomalies	25 (44.6%)
Hospitalized in a public institution	41 (73.2%)
Hospitalized in a private institution	15 (26.8%)
Vaginal birth	22 (39.3%)
Birth by cesarean section	34 (60.7%)
Complex gastroschisis	14 (25%)
Total days of hospital stay (median)	25 days (min 2, max 173)
Days at initiation of enteral feeding after the first surgery (median)	11 days (min. 2. Max. 40)
Days at initiation of enteral feeding after closure (median)	7 days (min.2, max. 40)
Completion of enteral feeding within 15 days after first surgery**	19 (33.93%)
Infectious complications	40 (71.4%)
Metabolic complications	44 (78.6%)
Pulmonary hypertension	9 (16.1%)
Acute kidney injury	13 (23.2%)
Pneumonia	9 (16.1%)
Post-surgical complications of surgery	13 (23.2%)
Mortality	12 (21.4%)
Age at death (median)	29 days (min 2, max 173 days)

* Of those born in Cali, 50% come from the east of the city. ** Of the deceased, one managed to complete the oral route

The median hospital stay was 25 days. Enteral feeding was initiated at a median of 11 days after the first surgery and at 7 days after closure. 33.93% of the patients achieved complete enteral feeding within 15 days after the first surgery. 71.4% (40 patients) presented infectious complications, and 79.6% (44 patients) had metabolic complications (Table 1). The mortality rate was 21.4% (12 patients.

Clinical Factors Significantly Associated with Mortality (Table 2):

Invasive mechanical ventilation for longer than 7 days (OR 7.94, 95% CI 1.55-40.73)

Presence of atelectasis (OR 4.76, 95% CI 1.24-18.3)

Pulmonary hypertension (OR 7.14, 95% CI 1.53-33.34)

Acute kidney injury (OR 8.87, 95% CI 2.11-37.23)

Pneumonia (OR 7.14, 95% CI 1.53-33.34)

Table 2: Clinical factors associated with the mortality.

VARIABLES	OR	95% CI	P
Intrauterine growth restriction	2.70	0.73 – 9.99	0.18
Maternal age < 19 years old	0.88	0.22 – 3.37	1.00
Origin outside of Cali*	0.76	0.21 – 2.73	0.67
Invasive mechanical ventilation > 7 days	7.94	1.55 – 40.73	0.00
No neuromuscular relaxation	1.52	0.57 – 4.00	0.06
Sepsis	8.35	0.99 – 70.5	0.04
Surgical site infection	0.27	0.03 – 2.36	0.42
Atelectasis	4.76	1.24 – 18.30	0.03
Pulmonary hypertension	7.14	1.53 – 33.34	0.01
Acute kidney injury**	8.87	2.11 – 37.23	0.00
Pneumonia	7.14	1.53 – 33.34	0.01

*Origin outside Cali refers to municipalities surrounding Cali
**Acute kidney injury refers to neonatal KDIGO classification [31]

Surgical Factors Significantly Associated with Mortality (Table 3):

Abdominal compartment syndrome (OR 13.67, 95% CI 2.68-69.71)
Staged closure (OR 9.17, 95% CI 1.09-77.24)
More than 2 surgical interventions (OR 7.8, 95% CI 1.80-33.77)
Failure to achieve closure (OR 30.71, 95% CI 3.11-303.55)
Intestinal ischemia (OR 43, 95% CI 4.39-421.59).

Table 3: Surgical factors associated with mortality

VARIABLES	OR	95% CI	P
Complex gastroschisis*	2.78	0.71 – 10.84	0.15
Abdominal compartment syndrome	13.67	2.68 – 69.71	0.00
First surgery after 3 hours of birth	1.44	0.40 – 5.20	0.57
Staged closure	9.17	1.09 – 77.24	0.02
Open abdomen with VAC system	0.41	0.10 – 1.67	0.09
Number of surgeries > 2	7.80	1.80 – 33.77	0.00
Impossibility to achieve closure	30.71	3.11 – 303.55	0.00
Post-surgical complications **	5.29	1.32 – 21.23	0.01
Intestinal ischemia	43	4.39 – 421.59	0.00

*Complex gastroschisis refers to those with atresia, perforation, volvulus, ischemia, or stenosis [1].

** Post-surgical complications such as stenosis, requirement of intestinal resection, obstruction, short-bowel syndrome, requirement of an ostomy, evisceration, intestinal ischemia, or intestinal perforation.

DISCUSSION

During the years 2018 - 2022, a total of 56 babies with a diagnosis of gastroschisis were identified from two institutions in the city of Cali, Colombia. These institutions report up to 60% of the city's births. The rate of live births with a diagnosis of gastroschisis was 1/1,000 live births, similar to that reported in another study at the HUV between 2000 and 2004 [12], and higher than the worldwide rate of 1.1 to 5.1 per 10,000 live births [6].

Regarding sociodemographic variables, multiple studies have found that the prevalence of gastroschisis is higher in mothers under 20 years of age [1], which is similar to our population, where the median maternal age was 19.5 years. Complex gastroschisis, defined as associated with congenital intestinal complications such as atresia, perforation, ischemia, necrosis, or volvulus, occurs in 17% of cases, close to the 25% found in our study.

Physiologically, the abdominal wall plays an important role in inspiration, exhalation, and clearance of the airways through coughing. The compliance of the abdominal wall is important for the activation of the diaphragm during inspiration; when it is affected, thoracic expansion is limited, and the respiratory muscles are weakened [17]. In our study, clinical factors significantly associated with mortality were centrally related to the respiratory system, including the requirement for invasive ventilation for more than 7 days, presence of atelectasis, pulmonary hypertension, and pneumonia. The abdominal wall is an integral part of the thorax, as its alterations affect respiratory mechanics and diaphragm function. The results of this study suggest the need to evaluate the relationship between abdominal wall defects and lung diseases and pulmonary hypoplasia, as described in other investigations [10, 17]. Consequently, it is necessary to evaluate these defects prenatally, using ultrasonographic measurements of the lung-to-thorax ratio, which was not reported in this investigation.

When gastroschisis surgical repair is performed, visceral-abdominal disproportion can result in respiratory compromise. This is because the reduction of the protruded organs results in high intra-abdominal pressures, leading to greater cephalic displacement of the diaphragm and limitation in its movement [18]. This results in decreased tidal volume, atelectasis, hypoxemia, and hypercarbia. Dimitriou et al. found that primary closure is associated with an approximately 40% reduction in respiratory compliance in the first 2 postoperative days [19].

After the clinical stabilization of the newborn, surgery is performed to return the eviscerated intestine to the abdominal cavity. This can be done in a single intervention (primary closure) or in stages. The ideal surgical method is debatable, but it takes into account the condition of the intestine and the disproportion between the viscera and the abdomen [6]. One of the most feared complications of this operation is abdominal compartment syndrome, which was significantly associated with higher mortality in our study. This condition presents compression in the mesenteric vessels and the

inferior vena cava, leading not only to lower cardiac output with subsequent renal and cerebral alterations but also to the risk of abdominal organ hypoperfusion [20]. Notably, intestinal ischemia was also significantly associated with higher mortality.

In most studies, primary closure is associated with better outcomes in New Zealand, Australia, and England [7, 9]. In Medellín, primary closure continues to be the most used procedure, with a percentage of complications similar to that of patients treated with staged closure, but with the advantage of a shorter time to initiation of enteral feedings. Likewise, it was found in Medellín that the use of a silo is not associated with infection of the surgical site but is associated with the same percentage of sepsis as in children treated with primary closure, as well as a more rapid return of intestinal function in patients treated with primary closure than those treated with a silo [10]. Importantly, staged closure was significantly associated with increased mortality in our study. This result can be explained by a selection bias, where patients with complicated gastroschisis and more severe clinical conditions are selected for staged closure. Additionally, abdominal pressure is not measured in the institutions before, during, and after surgery for all patients, which has been associated with better outcomes regardless of the type of surgery selected [20].

The prognosis and mortality in those born with gastroschisis depend extensively on the condition of the intestine at the time of birth [1]. Mortality due to gastroschisis in Mexico is variable; in a study by Valdez, a survival rate greater than 90% was reported, with the main causes of mortality being prematurity, neonatal sepsis, intestinal complications related to intestinal ischemia, acute renal failure, or multiple organ failure. In Mexico, gastroschisis represents the fourth leading cause of death in children under 5 years of age due to congenital malformations [5]. In the United States, a mortality rate of 3.4 to 9.3% has been found, which is lower than the 25% found in our study. Bidondo et al. report that the most frequent cause of mortality in their studies was compartment syndrome and abdominal complications requiring emergency rescue surgery [21], which was also significantly associated with mortality in our study. At the HUV for the period 2000-2004, 28 (96.6%) of 29 children with gastroschisis developed complications, with 82.1% developing sepsis, 53.6% electrolyte imbalance, 32.1% atelectasis, 21.4% pulmonary hypertension, 14.3% intestinal adhesions or flanges, 10.7% acute renal failure, 7.1% pneumonia, and one case each of liver failure, cholestasis, myocardial dysfunction, and compartment syndrome [12].

CONCLUSION

In this study, we identified that the incidence rate of gastroschisis in our setting is significantly higher than that reported in other regions, as evidenced by an increase from 5.15/10,000 births between 2000 to 2008 to 1/1,000 births in this investigation. This necessitates evaluating related factors, such as maternal age, which in our study had a median of 19.5 years and the potential relationship with environmental and pharmacological teratogens.

Additionally, the association of gastroschisis with pulmonary pathologies such as atelectasis, pneumonia, and pulmonary hypertension—which lead to tracheal intubation for more than seven days—highlights the need for implementing diagnostic measures of fetal lung conditions using ultrasonography. Surgical complications such as compartment syndrome, intestinal ischemia, and issues related to the closure of the defect were significantly associated with increased mortality. This underscores the importance of early detection of abdominal compartment syndrome after the closure of congenital abdominal wall defects, as it affects the functionality of the pulmonary, cardiovascular, renal, musculoskeletal, and central nervous systems.

Our institutions should emphasize the use of a quantifiable method to evaluate intra-abdominal pressure through a bladder catheter connected to a pressure transducer, which is recognized as a reliable method to prevent the development of abdominal compartment syndrome. These results highlight the need for continued research into factors that can reduce the incidence of this pathology and improve the survival rates of these babies in low- and middle-income regions.

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